ECONOMIC BURDEN ASSOCIATED WITH SJÖGREN'S DISEASE (SJD): FINDINGS FROM A SYSTEMATIC LITERATURE REVIEW (SLR)

Poster #EE220

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Background

- Sjögren's disease (SjD) is an autoimmune disease that is estimated to affect 0.3–26.1 per 100,000 individuals, more commonly affecting women than men.[1]
- SjD is typically characterized by dryness of the eyes and mouth (sicca symptoms); other symptoms include fatigue, musculoskeletal pain, and systemic features.[2]
- SjD treatments have not substantially improved symptom relief in recent decades.[2,3]
- Previous SLRs have not specifically addressed the economic burden of SjD.[4,5]

Objectives

- The objective of this SLR was to summarize the available evidence on the global economic burden of SjD with a focus on:
  - SjD severity (primary vs secondary SjD, EULAR Sjögren's Syndrome Disease Activity Index [ESSDAI] ≥5, and organ involvement)
  - Specific cost drivers (dental costs and healthcare resource utilization [HCRU]).

Methods

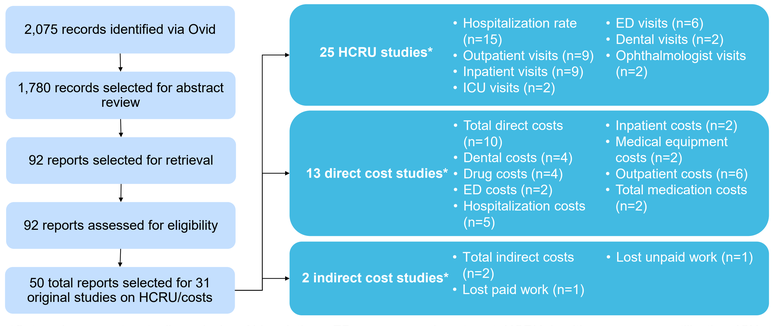
- This SLR followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines and was conducted in Ovid (databases included Embase, Medline, NHS EED, EconLit, and INAHTA) to identify cost and HCRU studies published 2012–2023.
- Population, Intervention, Comparison, Outcomes and Study (PICOS) criteria:
  - Population: primary or secondary SjD.
    - Subpopulations of interest: ESSDAI ≥5, ESSDAI <5, and EULAR Sjögren's Syndrome Patient Reported Index (ESSPRI) ≥5.
  - Intervention: no restriction.
  - Outcomes: resource use, direct costs, indirect costs, incremental cost-effectiveness ratio, economic model specifications, and data sources.
  - Study design: observational studies, piggyback trials, economic evaluations (e.g., cost-effectiveness analysis, cost-utility analysis, budget impact analysis).
- Editorials, commentaries, case reports, letters, and reviews were excluded, with any SLRs cross-checked for relevant studies.
- Conference proceedings (2020–2023) and bibliography searches (2012–2023) were used to validate search results and capture results not yet indexed via Ovid.

Results

Study characteristics

- Thirty-one studies were identified (Figure 1).
- Included studies were conducted in North America (39%), Europe (39%), Asia (13%), and the Middle East and South America (1%).
- Four studies included secondary SjD[6,7,8,9] and two studies included subpopulations of interest (ESSDAI and ESSPRI).[10,11]

Figure 1. HCRU/cost studies identified



*Categories are not mutually exclusive. Abbreviations: ED, emergency department; HCRU, healthcare resource utilization; ICU, intensive care unit.

Results (cont.)

Healthcare resource utilization

- Annual hospitalization rates were higher among patients with vs without SjD and varied between subgroups of SjD patients (Table 1).
- HCRU was higher among individuals with vs without SjD. More annual outpatient visits were reported with SjD vs without SjD (Figures 2a and 2b).

Table 1. Annual hospitalization rates

Study	Patient group (n)	Hospitalization rate	P-value
Hospitalization rates, SjD vs non-SjD			
Canada (1996–2008) hospitalization one year after diagnosis[12]	SjD (691)	0.29	<0.01*
	Non-SjD (6,910)	0.11	
Germany, (2018)[6]	Primary SjD (2,810)	0.32	NR
	Secondary SjD (3,750)	0.39	
	Matched controls (29,801)	0.21	
US (1995–2016; data per 100 patient-years)[13]	Primary SjD (160)	0.24	NR
	Comparator (466)	0.19	
US (2007; visits over 5 years)[14]	Primary SjD (277)	0.53	<0.05*
	Control (606)	0.40	
Sweden (2009†)[15]	Primary SjD (7,396)	0.50	<0.0001*
	General population (76,624)	0.30	
Hospitalization rates by SjD subgroups			
Germany (2018)[6]	Primary SjD (2,810)	0.32	NR
	Secondary SjD (3,750)	0.39	
Spain (year NR)[16]	SjD without renal disease (398)	0.15	0.004*
	SjD with renal disease (39)	0.33	
US (year NR)[10]	Median ESSPRI: 4	Recent SjD diagnosis (154)	0.14	NR
	Median ESSPRI: 5.3	Slower SjD progression (154)	0.17	
	Median ESSPRI: 6	Moderate severity/time since diagnosis (184)	0.22	
	Median ESSPRI: 6.5	Most severe SjD (132)	0.27	

Reported hospitalization rates are annual. *Statistical significance. †Patients diagnosed in 2009. Abbreviations: ESSPRI, EULAR Sjögren's Syndrome Patient Reported Index; NR, not reported; SjD, Sjögren's disease; US, United States.

Figure 2a. Number outpatient visits and dental visits in Sweden for patients diagnosed in 2009 (2015)[15]

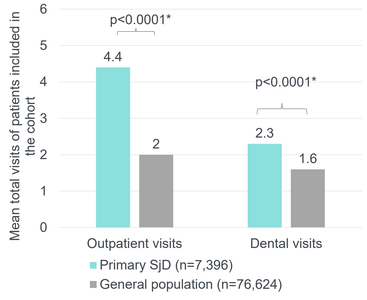
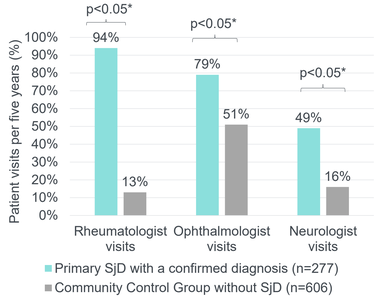


Figure 2b. Rheumatologist, ophthalmologist, and neurologist visits in the US over 5 years (2007)[14]



*Statistical significance. Abbreviations: SjD, Sjögren's disease; US, United States.

Healthcare resource utilization by subgroup

- More frequent emergency department visits were reported for SjD with vs without extra-glandular manifestations (mean 1.7 vs 1.3 per person per year [PPPY], respectively; no stats reported) and for secondary vs primary SjD (mean 0.15 vs 0.07 PPPY, respectively; no stats reported).[7,17]
- A high number of outpatient visits were reported in both primary and secondary SjD populations (mean 4.35 vs 3.06 PPPY, respectively; no statistics reported).[7]
- Patients with increasing ESSPRI scores were correlated with an increased number of healthcare provider visits (ESSRI 4.0, 5.3, 6.0, and 6.5, median 3, 4, 4, and 5 visits).[10]
- Additional subgroup data are provided in the supplementary materials.

Direct and indirect costs

- Direct costs were higher among patients with SjD vs non-SjD matched/community controls. In Sweden, hospitalizations were greater cost drivers than outpatient or drug costs.[10,16] In the United Kingdom (UK), the cost of healthcare professionals was the main cost driver over hospital stays, diagnostic tests, or drug therapy costs.[18]

Results (cont.)

- Total direct costs and dental costs were higher among those with vs without SjD (Table 2).
- Total direct costs between subgroups of patients with SjD were either not statistically different or not reported.
- In a United States (US) study, inpatient costs and outpatient costs were the leading cost drivers for patients with SjD and ESSDAI scores ≥5.[11]
- In the UK, indirect costs were higher with SjD vs controls (p-values: total indirect costs, <0.001; paid work, <0.001; unpaid work, <0.05).[19]

Table 2. Direct and indirect costs

Study/cost type	Patient group (n)	Cost (currency)	P-value
Total direct costs, SjD vs non-SjD			
Swedish study (2016)[20]	Primary SjD (8,884)	51,384 (SEK)	<0.0001*
	Non-SjD matched controls (88,233)	28,497 (SEK)	
UK study (2004–2005)[18]	Primary SjD female (129)	2,188 (GBP)	*†
	Non-SjD matched controls female (92)	949 (GBP)	
US study (2013)[17]	Primary SjD (12,717)	10,729 (USD)	NR
	Non-SjD matched controls (12,717)	6,534 (USD)	
Dental costs (direct), SjD vs non-SjD			
Swedish study (2016)[15]	Primary SjD (8,884)	3,724 (SEK)	NS
	Non-SjD matched controls (88,233)	2,724 (SEK)	
UK study (2004–2005)[18]	Primary SjD female (129)	452 (GBP)	*†
	Non-SjD matched controls female (92)	302 (GBP)	
US study (year 2007)[14]	Primary SjD confirmed diagnosis (277)	1,473 (USD)	<0.05
	Non-SjD community controls (606)	504 (USD)	
Direct costs by SjD subgroup			
Mean healthcare costs (France, 2018)[21]	Primary SjD (23,152)	9,618 (EUR)	NR
	SjD and autoimmune disease (15,462)	13,271 (EUR)	
Mean total direct costs (US, 2013)[17]	Primary SjD glandular disease only (4,997)	4,878 (USD)	NR
	Primary SjD extra-glandular disease manifestations (7,720)	14,387 (USD)	
Annual mean total direct costs (US, year NR)[11]	ESSDAI <5 (194)	11,165 (USD)	NS
	ESSDAI ≥5 (19)	15,656 (USD)	
Mean total direct costs (US, 2020)[22]	SjD, no interstitial lung disease (4,075)	3,369 (USD)	NR
	SjD, interstitial lung disease (815)	7,960 (USD)	
Indirect costs, SjD vs non-SjD			
UK study (2008)[19] Total	Primary SjD (84)	12,362 (GBP)	<0.001*
	Non-SjD community controls (96)	1,057 (GBP)	
UK study (2008)[19] Paid work	Primary SjD (84)	10,840 (GBP)	<0.001*
	Non-SjD community controls (96)	705 (GBP)	
UK study (2008)[19] Unpaid work	Primary SjD (84)	1,376 (GBP)	<0.05*
	Non-SjD community controls (96)	330 (GBP)	
Chinese study (2016)[23]	SjD with dry eye (34)	828 (CNY)	0.017*
	Non-SjD with dry eye (30)	487 (CNY)	

*Statistical significance. †Alpha value not reported. Abbreviations: CNY, Chinese Yuan; ESSDAI, EULAR Sjögren's Syndrome Disease Activity Index; EUR, Euro; GBP, Great British Pound; NR, not reported; NS, not significant; SEK, Swedish Krona; SjD, Sjögren's disease; UK, United Kingdom; US, United States; USD, United States Dollars.

Conclusions

- This is the first global SLR investigating the economic burden of SjD. Overall, direct costs and HCRU were higher among patients with SjD vs healthy controls, and dental costs were higher among patients with SjD vs non-SjD matched controls.
- This SLR was limited by the availability of data: in particular, there was a lack of subgroup data on patients with ESSDAI and/or ESSPRI scores >5 and limited data comparing patients with primary vs secondary SjD.
- Our findings suggest that patients with SjD have a substantial economic burden, which increases with disease and symptom severity; however, more research is required to identify which SjD characteristics are associated with higher costs/HCRU.

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Disclosures

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